ENVIRONMENTAL INFLUENCES RELATED TO THE AETIOLOGY OF CONGENITAL DISLOCATION OF THE HIP

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Congenital dislocation of the hip has been recognized for many centuries and has given rise to a literature so extensive that a comprehensive review cannot be attempted here. The condition was described and named by Hippocrates, who suggested uterine pressure and birth trauma as possible causes.

Paré published a more detailed description of the defect and recognized its occasional hereditary nature. Extensive clinical and pathological studies were made during the 18th and 19th centuries but failed to suggest any more precise explanation than that of a diathesis, manifestations of which depended on various environmental influences. The only evidence of environmental hazard related to breech presentation, which was believed to predispose to dislocation either by foetal deformation or obstetric trauma. An excellent review by Sainton (1893) covers the early history.

Vogel (1905) made the first detailed study of the influence of heredity and of the uterine environment, finding that an affected relative could be remembered in about a third of the families studied, and that breech presentation and other obstetric difficulties were unduly common in the births of affected children. Le Damany (1912, 1914) made a major advance in distinguishing cases developing as a consequence of gross abnormalities, such as spina bifida, from the majority which he regarded as merely one end of a continuously variable distribution of hip stability. In his elaboration of Darwin's view of malformations as extreme varieties, he may claim precedence in applying the concepts of quasicontinuous variation and polymorphism to man. His views on congenital dislocation of the hip are strictly analogous to contemporary views of mental deficiency, which may either result from metabolic abnormality or local disease, or be merely the lower end of a continuous distribution.

Unfortunately, these two papers appeared at a time when human genetics was passing through a

phase of rapid change. Effort was concentrated in attempting to apply Mendel's concepts to the inheritance of human characteristics and diseases. and workers tended to think in terms of single gene substitutions. Environmental influences were largely ignored and genetical hypotheses of increasing complexity were formulated in an attempt to satisfy observational data. Isigkeit (1928) for example, suggested that congenital dislocation of the hip was determined by two sex-linked dominant genes (one of which was presumed to be semi-lethal when recessive) and a pair of recessive autosomal genes. Faber (1937) concluded, from radiographic measurements of relatives of affected persons, that a dysplasia of the hip, defined in terms of these measurements, is inherited as a Mendelian dominant of incomplete penetrance. This view, strongly supported by Hart (1947), received wide acceptance and is considered by some to be so well established as to form the basis for eugenic counselling and even for the sterilization of so-called latent carriers. Faber's interpretation of his data, however, is open to doubt because the measurements show no evidence of bimodality. In fact, the data appear to show little more than that in about half the cases the measurements were below the average of the whole series. The limitations of the interpretation may be illustrated by considering a hypothetical disease predisposed to by tallness. If measurements of relatives of a few affected persons were made, it would be possible to define tallness in such a way that it would appear to be transmitted as a Mendelian dominant of incomplete penetrance.

A careful twin study by Idelberger (1951) may help to re-orientate opinion in a more promising direction. An examination of 138 living twin pairs showed that concordance occurred in less than half the monozygous pairs, and that among dizygous pairs the twin of an affected member was no more likely to have the defect than any other sibling of the same sex. Although some of Idelberger's conclusions are not acceptable, his observations are invaluable and seem to be consistent with the view of earlier workers that a number of factors, both genetical and environmental, must be implicated. It seems doubtful whether the hereditary background will be understood until more is known of environmental influences, and the present work was planned to explore these factors.

MATERIAL

An attempt was made to assemble a comprehensive series of cases of congenital dislocation of the hip occurring in children born in the years 1942-52. This period was chosen because an estimate of the maternal age and parity distribution of all Birmingham births for these years was already available from a previous investigation (Smith and Record, 1955). Inquiry at all hospitals in Birmingham indicated that treatment for congenital dislocation of the hip was undertaken at only the Children's Hospital and the Royal Orthopaedic Hospital. The diagnostic indexes of these hospitals provided the names and addresses of 202 patients whose dates of birth came within the specified period (Table I). Sixteen lived more than 12 miles away and were excluded from the survey. An attempt was made to visit the homes of the remaining 186 patients. In nineteen cases the family had moved and could not be traced. The rest of the series (167 cases) were visited and the mothers were interviewed. These cases provide the data on which most of the analysis is based, but the whole series of 186 propositi has been used to examine seasonal variation in incidence, the date of birth being known for all the children. For those parts of the analysis which required knowledge of the related population, attention was restricted to propositi whose parents resided in Birmingham when the affected children were born; patients whose parents lived outside the city were excluded since they came from an area

TABLE I

CASES OF CONGENITAL DISLOCATION OF THE HIP BORN
IN THE PERIOD 1942-52

Number of Affected	Domicile at 7	Time of Birth	77-4-1
Number of Affected	Birmingham	Elsewhere	Total
In Hospital Records Excluded from Survey* In Survey Untraced Whose Parent was Interviewed	148 148 12 136	54 16 38 7	202 16 186 19

^{*} Because domicile was more than 12 miles from Birmingham,

with no precise boundaries. The estimate of incidence, therefore, is based on the 148 Birmingham cases, and the examination of the influence of maternal age and parity is restricted to the 136 who were visited.

RESULTS

INCIDENCE

The 148 cases among Birmingham children born in the years 1942-52 may be related to the total number of live births (226,038) computed from data published annually in the Registrar General's Statistical Reviews. This gives an incidence of 0.65 per 1,000 live births (0.2 and 1.1 per 1,000 for males and females respectively). This is lower than most published estimates, but it is well known that incidence shows very marked regional variations. Among the Lapps, incidence is said to be about 4 per cent. (Getz, 1955), and considerably higher values have been reported in some Lapp villages (Allison, 1956), and in Red Indian settlements (Steinberg, 1957). The condition is rare among Negroes and almost unknown among the Sudanese. It is possible that most recorded estimates come from areas of high prevalence where the condition is more likely to be studied because of its more obvious social and economic importance.

It may be advisable, however, to consider the reliability of the present estimate. Ascertainment may have been incomplete for the following reasons:

- (1) Some affected infants may have died before the abnormality was detected. There is undoubtedly a relationship between congenital dislocation of the hip and several conditions such as breech presentation and low birth weight which are associated with raised perinatal mortality.
- (2) Some children may have moved outside the Birmingham area before the defect was recognized. It seems likely that this was rare. Among the 167 affected in the series who were traced there was only one whose parents had lived outside Birmingham when the child was born and had later moved into the city. It is reasonable to suppose, therefore, that not more than one or two cases were lost by emigration, since during the period of the survey more people were coming into the city to live than were leaving it.
- (3) At the time when the present investigation was begun the condition may not have been diagnosed in some children born in the later years of the period. We believe that such losses are very small since the survey was not started until the youngest children had attained the age of 3 years. 85 per cent. of the

cases in the series had been diagnosed by the age of 2 years, and only 3 per cent. were diagnosed after the third birthday.

(4) Some cases in older children may not have been diagnosed, or the diagnosis may have been made too late for treatment to be attempted. We have no knowledge of the number of these cases, but in view of the efficiency of the Child Welfare and School Medical Services in Birmingham it seems certain that it is small.

It seems reasonable to conclude that the figure of 0.65 per 1,000 births for the incidence of congenital dislocation of the hip in Birmingham may be slightly below the true value, but is probably acceptable as an estimate of incidence among children who survived infancy.

CHARACTERISTICS OF PROPOSITI

- (a) Sex Ratio.—The complete series consisted of 27 males and 159 females. The proportion of males (14·5 per cent.) is in close agreement with the sex ratio (14·9 per cent.) estimated by Poli (1937) on 37,503 cases reported in the literature. The higher incidence in females was considered by le Damany (1912) to be due to sex differences in the structure of the pelvic bones and particularly in the inclination of the acetabula. This view was supported by Dega (1933), who found sex differences in the shape of the foetal pelvis, even as early as the third month.
- (b) Side Affected.—In two cases details of the affected side were not available. Among the remaining 184, the condition was bilateral in 47 (25.5 per cent.) and involved the left hip only in 101 and the right hip only in 36. The number of affected hips in the series was, therefore, 231, of which 148 (64.1 per cent.) were left-sided. Nearly all published series show a higher incidence on the left side than on the right, but the discrepancy is not usually as marked as in the present series. There appears to be little difference between males and females in the relative frequency of the double lesion (Table II).

TABLE II
AFFECTED SIDE ACCORDING TO SEX

Affected	М	lales	Fer	nales*	7	otal
Side	No.	Per cent.	No.	Per cent.	No.	Per cent.
Left only Right only Both	15 4 8	55·6 14·8 29·6	86 32 39	54·8 20·4 24·8	101 36 47	54·9 19·6 25·5
Total	27	100	157	100	184	100

^{*} Two cases for whom the affected side was not stated have been excluded.

There is even less difference between the sexes in the frequency of involvement of the left hip relative to the right; the 27 male propositi had 35 affected hips, of which 23 (66 per cent.) were left-sided; the 159 female propositi had 196 dislocations, of which 125 (64 per cent.) were left-sided.

(c) Associated Defects.—Details of associated abnormalities were obtained during the home visit and are based, therefore, on the 167 propositi who were traced. Defects, details of which are given in Table III, were present in 26 (16 per cent.). Although no strictly comparable rate is available for the general population, there seems little doubt that this incidence is very high, and is the more remarkable when it is recalled that the series consisted of children who survived infancy and would, therefore, be unlikely to exhibit the more serious malformations. Grundy and Lewis-Faning (1957), in an extensive survey of infants born in 1952 in fifteen areas of England and Wales, found a malformation rate among live births of 2.9 per cent., and onequarter of these infants died before the first birthday. There are many reports in the literature of the frequent association of congenital dislocation of the hip with other deformities, but an incidence as high as 16 per cent. is rare. Another interesting feature is that three of the 26 propositi with associated defects were twins. The defects were hare-lip and cleft palate, torticollis, and diaphragmatic hernia. Since the whole series contained only four twins, the risk of associated defects appears to be much greater for twins than for single births, but this view is not supported by the observations of Idelberger (1951).

TABLE III
ASSOCIATED DEFECTS OCCURRING AMONG
167 PROPOSITI

Type of Defect	Diagnosis	No.	Per cent.		
Nervous	Spina Bifida and Hydrocepl Spastic Paralysis of Hand Spastic Paralysis and Menta Mental Defect Amyotonia	Defect	1 2 2 1 1	}	4.2
Musculo- Skeletal	Diaphragmatic Hernia Other Herniae Torticollis Dislocation of Patellae Genu Valgum Talipes Pes Planus	· ··	1 3 2 1 1 8	}	10-2
Other	Hare-lip and Cleft Palate . Hypospadias		1 1	}	1.2

(d) Birth Weight.—The weights of the propositi at birth, which were obtained from the mother, were available for 161 of the 167 cases visited. The

distribution of weight was compared with that derived from a survey of all births in Birmingham in 1947 (assembled by Gibson and McKeown, 1950), from which all stillbirths and infant deaths have been excluded. Because of the variation of birth weight with sex and birth rank it was necessary to standardize the controls to make them comparable in respect of these two variables with the series of propositi. When the percentage distribution of affected children is compared with that of the standardized controls, there is an excess of propositi weighing less than 7 lb. and a deficiency of those weighing 9 lb. and over (Fig. 1). The difference between the two distributions is also shown by a comparison of the means. The mean birth weight of affected children was 6.81 lb. and that of the standardized controls 7.36 lb. (difference $0.55 \pm$ 0.09).

This difference, although apparently slight, implies a very considerable increase in risk to small babies. It may be shown that, if birth weight were normally distributed and the affected and the controls had the same standard deviation of about 1 lb., then a difference in birth weight of d lb. would make a difference in relative liability of about $e^{0.55d}$; that is,

there would be a doubling in incidence associated with each 1·3 lb. deficiency in birth weight. This does not, of course, imply that influencing birth weight would necessarily influence liability to dislocation, as babies prone to dislocation may form a distinct sup-group.

It may be objected that a comparison between the two series is not valid because the weights of propositi were obtained by interrogation of the mother, and the control series was based on observed weights. We have evidence from another inquiry that the great majority of mothers accurately remember the birth weights of their children. The weights of all infants born in a neighbouring town (Smethwick) during one year was recorded. Two years later, the mothers were visited and asked what the birth weight was. 907 mothers were interviewed; a correct statement was obtained from 536 (59 per cent.) and a further 221 (24 per cent.) were not more than 4 oz. in error. Errors greater than a pound were recorded for only eighteen (2 per cent.). The errors of those who understated the weight (193) were very nearly cancelled by those who overstated it (178), and the net effect on the mean weight of the whole series was an underestimation of only 0.005 lb.

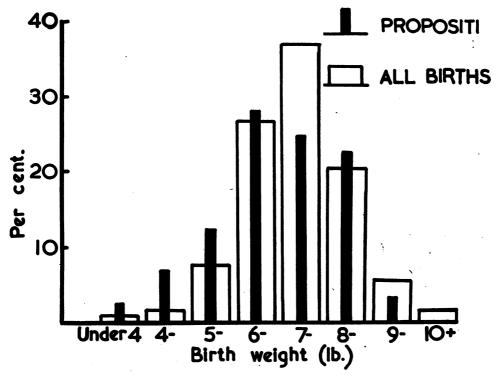


Fig. 1.—Distribution according to birth weight compared with that of all Birmingham births in 1947 (first-year survivors) of the same sex and parity distribution,

There seems to be little doubt, therefore, that infants with congenital dislocation of the hip were lighter than average at birth. This result leads one to inquire whether they were born earlier than usual. Our data are hardly adequate to answer this question with certainty. Mothers were interrogated about the duration of the pregnancy, but could not, of course, be expected to remember the date of the preceding menstrual period, and rather approximate answers such as "full time" or "about the expected date" were often received. Of 165 women who attempted a reply, 140 (85 per cent.) gave histories of pregnancies lasting at least 39 weeks. The corresponding figure derived from the 1947 Birmingham survey (after excluding stillbirths and infant deaths) was 82 per cent., but this was a more reliable estimate, since durations of pregnancy were calculated from the last menstrual period. This rather inadequate evidence suggests that there is nothing unusual about the gestation periods of propositi. This conclusion is not consistent with results reported by Vogel (1905), who found that 24 per cent. of his cases were born at least a fortnight early.

ASSOCIATION WITH MATERNAL AGE AND BIRTH RANK

In order to examine the variation in incidence with maternal age and birth rank in circumstances where the use of the Greenwood-Yule method is inappropriate (McKeown and Record, 1956), we require to know the age and birth rank of affected children and of the population of births from which the series was assembled. The analysis which follows is based on the 136 propositi whose parents lived in Birmingham at the time of the birth and were later traced, and a control group of 1,156 births (live and still) sampled from the 231,619 infants born to Birmingham residents during the years 1942–52. When the data are set out in a maternal age and birth rank contingency table (Table IV), the ratio of the number of propositi to the number of controls in

each cell can readily be converted into an estimate of incidence by multiplying by the ratio of the total number of controls (1,156) to the total number of births (231,619). (The total incidence thus calculated differs slightly from that given in the previous section because the related population includes stillbirths, and because it ignores the twelve Birmingham cases which were not traced.)

The most prominent feature of the data is the relatively large proportion of affected first-born (50.4 per cent. compared with 34.3 per cent. in the control series). The raised incidence among first-born occurs in all the maternal age groups. The liability of first-born to dislocation appears to be about double that of second and third-born. A primogeniture effect of this magnitude does not appear to have been previously recorded for any malformation.

The influence of maternal age is less marked. Incidence among first born rises with increasing maternal age, but for second and third children there seems to be no association. In the higher birth ranks, numbers in the younger age groups are too small to show a trend, but incidence is high among children born to women aged 35 and over.

It may be asked whether the increased risk among first-born can be explained by the greater incidence among children of low birth weight, since first children generally weigh less at birth than later children. We have explored this problem by redistributing the 1947 series of Birmingham births to give the same weight distribution (taking account of sex) as the series of propositi. The method is simple, being merely a standardization procedure, but the calculation is rather too lengthy to reproduce here. Its effect was to raise the percentage of first-born in the 1947 series from $40 \cdot 1$ to $42 \cdot 9$, *i.e.* an increase of $2 \cdot 8$, which is small compared with the observed difference of $16 \cdot 1$ ($50 \cdot 4 - 34 \cdot 3$ per cent.). It is concluded that the increased frequency of congenital

	٦	Table IV				
INCIDENCE ACCORDING	то	MATERNAL	AGE	AND	BIRTH	RANK

Birth Rank	Maternal Age (yrs)			Under 25	25–29	30–34	35 and Over	All Ages
1	No. of Propositi	::	::	30 200 0·75	24 126 0·95	12 46 1·30	2 25 (0·40)	68 397 0·85
2 and 3	No. of Propositi		::	12 125 0·48	13 187 0·35	14 159 0·44	5 79 0·32	44 550 0·40
4 and Over	No. of Propositi	::	::	0 2 —	3 42 0·36	3 68 0·22	18 97 0·93	24 209 0·57
All Birth Ranks	No. of Propositi	•••		42 327 0·64	40 355 0·56	29 273 0·53	25 201 0·62	136 1,156 0·59

dislocation of the hip among first-born cannot be accounted for by low birth weight.

There is no evidence in the Age-Parity Table of any association with social class. Variation from this source would be expected to produce a diagonal stratification of incidence in Table IV, since births in poor circumstances are represented mainly towards the left lower corner and those among the well-to-do towards the right upper corner.

MATERNAL FERTILITY

The slight association between incidence and advancing maternal age suggests the possibility of some impairment of fertility of mothers of propositi. Data relating to mothers of controls suitable for an examination of this problem were collected for a previous inquiry (McKeown, MacMahon, and Record, 1951a), but unfortunately are complete only for births before 1948. In order to make the series of affected children comparable with this control group, all propositi born after 1947 have been excluded, leaving 87 mothers for consideration. Date of marriage was recorded for 84 mothers. The mean age at marriage was 22.8 years for these women and 22.5 years for mothers of the controls. The difference seems to be too small to account for any difference in fertility between the two groups.

The interval preceding the conception of propositi was estimated for first-born from the date of marriage, pre-marital conceptions being excluded. For later-born propositi the interval was calculated from the date of the preceding birth. Data relating to 78 mothers of propositi were available and the mean fallow period was 38.5 months. The corresponding figure for the mothers of the controls standardized to the same parity distribution was 31.1 months. A measure of the mean relative fertility of the mothers of affected children can be obtained by calculating the ratio of the reciprocals of the two means. This gives a value of 0.81.

Fallow periods preceding conceptions of children born before propositi were calculated in the same way. Mothers of affected had had 53 earlier conceptions and the mean preceding fallow period was 22.8 months. The fallow period for mothers of controls standardized for parity was 18.5 months. The mean relative fertility of mothers of affected was again 0.81.

It appears, therefore, that women who give birth to children with congenital dislocation of the hip are, on the whole, slightly less fertile than other mothers. This conclusion is consistent with the finding that incidence among children of a given birth rank tends to rise with increasing maternal age. It does not, of course, permit one to decide whether

mothers of affected children are older because they are less fertile than other mothers or are less fertile because they are older.

Evidence bearing on this problem may be obtained by investigating fertility after the birth of an affected child. Some caution in interpreting the pattern of subsequent reproduction is necessary because of the possibility that it may be influenced by eugenic considerations, but if one may judge from studies of the more lethal malformations, such as those involving the central nervous system, or of the socially less acceptable defects such as mongolism, it would appear that parents are rarely deterred from further reproduction by the possibility that a later child may also be affected.

Mean fallow period is not an efficient index of subsequent fertility; a more useful measure is the number of children born after the propositus in a given time. The control series used above, based on interrogation of mothers in 1948, is not suitable for this investigation because of the relatively short period between the births of propositi and the home visit. Instead we have used a control group which was assembled for comparison with a series of mongols (Smith and Record, 1955). This group consisted of 139 mothers of children born in the years 1942-52; information on the number of subsequent children born by the end of 1953 was obtained by home visits. It was selected from the original control series of 1,156 births on the basis of maternal age to match so far as possible the mothers of the mongols. It contains, therefore, an undue proportion of older mothers, but this difficulty can be overcome by standardizing for age. Comparison between these mothers and the mothers of children with congenital dislocation of the hip shows a marked difference in fertility (Table V, overleaf). After standardizing to make the two groups comparable in respect of year of birth of propositi, the mean number of subsequent viable pregnancies was 0.34 per mother for the affected series and 0.43 for the controls. The ratio of these two values (0.79)approximates closely to the figures, obtained earlier, of relative fertility before the birth of propositi.

It seems reasonable to conclude that mothers of children with congenital dislocation of the hip show some impairment of fertility both before and after the birth of the affected child.

DISORDERS OF PREGNANCY

Retrospective inquiries into the health of women during pregnancies which resulted in malformed infants are of limited value since it is likely that mothers more readily recall illnesses in pregnancy when the child is abnormal. We have attempted to

	Unde		25		30		35		40 and		All	Δ ges
Maternal Age (yrs) at Birth of Propositus	Affected	Con- trols										
No. of Mothers No. of Subsequent Viable Pregnancies by	47	14	50	21	38	29	24	39	8	36	167	139
End of 1953	32	10	31	9	4	16	5	14	0	2	72	51
Mean No. of Subsequent Viable Pregnancies per Mother*	0.68	0.73	0.60	0.31	0-11	0.56	0.22	0 · 40	0.00	0.05	0.34	0.43

Table V
FREQUENCY OF PREGNANCIES WHICH OCCURRED AFTER BIRTHS OF PROPOSITI

overcome this difficulty by comparing the present series, not with a sample of births from the general population, but with a Birmingham series of cases of pyloric stenosis which was the subject of an earlier retrospective inquiry (MacMahon, Record, and McKeown, 1951). Pyloric stenosis seemed to be a suitable control since there is evidence that it is dependent much more on post-natal circumstances than on the pre-natal environment (McKeown, MacMahon, and Record, 1951b, 1952). The two series show some differences in the frequency of antenatal disorders (Table VI). This may be due to the small size of the hip series and to the fact that the two groups of mothers were interrogated by different observers. Nevertheless, there is suggestive evidence of an increased liability to toxaemia among mothers of children with hip dislocations. There seems to be no undue prevalence of bacterial or viral disease among the mothers; no instance of rubella was recorded.

TABLE VI
FREQUENCY OF ANTENATAL DISORDERS AMONG
PATIENTS WITH CONGENITAL DISLOCATION OF THE
HIP OR PYLORIC STENOSIS

Antenatal Condition	Cong Dislo	ents with enital cation e Hip		ents with Stenosis
	No.	Per cent.	No.	Per cent.
Excessive Vomiting Threatened Abortion Toxaemia Falls and Other Accidents Bacterial and Viral Disease	15 5 12 8 6	9·0 3·0 7·2 4·8 3·6	31 19 23 37 30	6·4 3·9 4·7 7·6 6·1

COMPLICATIONS OF DELIVERY

The history of the confinement obtained by retrospective questioning of the mother might also be expected to show some bias, but we believe that, if consideration is restricted to well-defined conditions of which the mother would certainly be told,

reliable records can be obtained. It is likely, therefore, that the incidence of the complications listed in Table VII, with the possible exception of "long or difficult labour", are reasonably accurate. Assessment of these results has been facilitated by the publication of data relating to all Birmingham births since 1949 in the Annual Reports of the Medical Office of Health. We used for comparison a single year (1950), but we satisfied ourselves by reference to later reports that this was typical and that the rates did not show a downward secular trend.

TABLE VII
INCIDENCE OF COMPLICATIONS OF BIRTH AMONG
167 MOTHERS OF PROPOSITI AND AMONG ALL
BIRMINGHAM BIRTHS IN 1950

Complications of Birth		ers of positi	All Birmingham Live Births
Complications of Birth	No.	Per cent.	in 1950* (Per cent.)
Breech Delivery Long or Difficult Labour	27	16.2	3.0
(Vertex Presentation) Twin Delivery Placenta Praevia	20 4 2	12·0 2·4 1·2	2·7 0·3†
Method of Delivery: Forceps (Vertex Presentation) Caesarean Section	10 6	6·0 3·6	3·6 2·1

^{*} Derived from data published in the Report of the Medical Officer of Health of Birmingham, 1950.

The results confirm the previously reported association of congenital dislocation of the hip with breech delivery. The association is particularly marked in the case of males: excluding cases of Caesarean section, there were 23 males of whom ten (43 per cent.) were delivered by the breech. Of the 138 female births, seventeen (12 per cent.) were breech deliveries. The difference between these proportions is much too large to be reasonably attributed to chance. The difference between the sexes is particularly marked for propositi of Birth ranks

^{*} Standardized to the distribution by year of birth of the combined series.

[†] Standardized to the distribution by year of birth and by maternal age of the combined series.

[†] Based on Birmingham live births in the years 1946-51.

2 and over (Table VIII). Of the thirteen male births in this group, seven (54 per cent.) were breech deliveries; the corresponding figure for females was three in 73 (4 per cent.).

TABLE VIII
FREQUENCY OF BREECH DELIVERY ACCORDING TO SEX
AND BIRTH RANK

Birth Rank	Sex	No. of Breech Deliveries	No. of Cephalic Deliveries	Frequency of Breech Delivery (Per cent.)	χ ₂ ,	p	
1	Male	3	7	30	0·03	0.9	
•	Female	14	51	22	0.03	0.9	
2 and	Male	7	6	54	21.95	<0.0001	
Over	Female	3	70	4	21.93	< 0.0001	
Total	Male	10	13	43	11.56	< 0.001	
Iotai	Female	17	121	12	11.30	< 0.001	

Although no comparable figure is available for the general population, the incidence of long or difficult labour (12 per cent.) seems to be higher than one would expect. This impression is supported by the raised frequency of forceps delivery, but assessment of these results is difficult because of the higher proportion of first births among propositi than in the general population.

The number of twin deliveries does not differ appreciably from the expected value. The same conclusion was reported by Idelberger (1951). The incidence of placenta praevia appears to be greater than usual, but numbers are too small to establish a definite association.

There was no obvious difference between bilateral and unilateral cases in the frequency of complications of delivery.

SEASON OF BIRTH

Dates of birth were obtained from hospital records and were therefore available for the whole series of 186 cases. When the seasonal distribution of these cases is compared with that of all maternities in England and Wales for the same years, the number of propositi born in the first and last quarters of the year is greater than expected, and there is a corresponding deficiency of cases in the second and third quarters. The differences are particularly marked in the first and third quarters (Table IX).

In attempting to account for these discrepancies, we may first consider the possibility that the seasonal distribution of births in Birmingham differs from that of the rest of England and Wales. There is evidence from a previous inquiry (McKeown and Record, 1951) that this is not so; for the years

1940-47 the percentage distributions of Birmingham births in the first, second, third, and fourth quarters were respectively 24·9, 25·9, 24·7, and 24·5. These figures do not differ appreciably from the "expected" percentages shown in Table IX.

TABLE IX

NUMBER OF CASES OF CONGENITAL DISLOCATION OF
THE HIP ACCORDING TO MONTH OF BIRTH

Month of	No. of	Percentage	Distribution	Difference O - E	
Birth	Propositi	Observed (O)	Expected* (E)	(per cent.)	
January February March	20 21 22	33.9	25·2	$+8\cdot7\pm3\cdot2$	
April May June	8 16 15	21.0	26.0	$-5\cdot0\pm3\cdot2$	
July August September	11 10 12	17.7	25.0	$-7\cdot3\pm3\cdot2$	
October November December	14 13 24	27 · 4	23 · 8	+3·6±3·1	
Total	186	100	100		

^{*} Based on all maternities in England and Wales, 1942-52.

The second possibility is that, although all births do not show any marked seasonal fluctuation in incidence, first births and later births separately may do so. A series which contained an unusual proportion of first born would then show a seasonal trend. National statistics do not permit a direct examination of this point because there are no published data on the monthly or quarterly incidence of births in relation to parity. There are, however, statistics relating season of birth to maternal age (Annual Reports of the Registrar-General, Pt. II), and these provide useful information, since it may be reasonably assumed that the great majority of mothers under 20 are primiparae and that most women aged 40 and over at the time of the birth are multiparae. These two groups show surprisingly little difference in the monthly distribution of births; in 1950 the proportion of births which occurred in the 6 months from April to September was 50.4 per cent. for women under 20 and 50.3 per cent. for women of 40 and over. It appears, then, that the association of incidence of congenital dislocation of the hip with season of birth is not due to its association with primogeniture. The same conclusion is reached if first and later born propositi are examined separately for evidence of seasonal variation. After excluding nineteen cases of unknown birth rank, there were eighty first born, 32 of whom were born in the period April-September. Of the 87 later born, 34 were born in the same period.

There seems to be little doubt, therefore, that there is some relationship between the incidence of congenital dislocation of the hip and season of birth. This might be due to one or more of a large number of factors, but we here consider only the simplest and most obvious one-temperature. Mean air temperatures, based on Birmingham records for the last 60 years, were obtained from the Annual Reports of the Medical Officer of Health. The relative incidence of congenital dislocation of the hip was calculated for each month by dividing the number of cases born in the month by the number which would have been expected if the condition were independent of season. The trend of these values, shown in Table X, is not perfectly smooth because of the small size of the series, but if April (where the incidence appears to be understated) is excluded, there is a steady gradation from the high values of the winter months to the low figures of July and August. There is obviously a marked negative association between relative incidence and air temperature, but it cannot be decided by inspection of the data whether incidence is correlated most strongly with the temperature of the same month or with that of preceding or succeeding months. This was examined by calculating co-efficients of correlation between relative incidence and the mean air temperature of the same month and of each of the two earlier and of the two later months. For example, the incidence in January was related in turn to temperatures in November, December, January, February, and March. (It may be objected that the relationship between incidence and temperature is not linear, and in fact, the regression is much better expressed by a quadratic curve. The use of the correlation coefficient in these circumstances is justified by the fact that it is used here only

TABLE X

RELATIVE INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP AND MEAN AIR TEMPERATURE ACCORDING TO MONTH OF BIRTH

nth o	f Birth		Relative Incidence	Mean Air Temperature (°F.)
::			1 · 28 1 · 43 1 · 33	38·8 39·2 42·0
::	::		0·50 0·96 0·95	46·3 52·2 57·4
::	::	::	0·69 6·66 0·79	60·8 60·1 56·0
::	::	::	0·94 0·91 1·59	49·7 43·0 39·9
				Incidence

as a comparative index.) The results (Table XI) suggest that incidence in any month is related more closely to the temperature of the same month than to that of the preceding or subsequent months. It is possible, however, that the abnormally low incidence in April may have a disproportionate influence on the correlations and it seemed advisable to repeat the calculations after excluding cases born in this month. This procedure gives slightly closer correlations, and the temperature in the month before birth seems to be at least as important as that of the month of birth.

TABLE XI
CORRELATION BETWEEN MONTHLY RELATIVE
INCIDENCE AND MEAN AIR TEMPERATURE OF
EARLIER, SAME, AND LATER MONTHS

Month of Temperature Record relative to			Coefficient of Correlation			
Month of		'	All	Cases	Excluding Cases Born in April	
2 Months Earlier				0.35	- 0.63	
1 Month Earlier			_	0.64	- 0.89	
Same Month]	_	0.72	- 0.87	
1 Month Later			_	0.65	- 0.67	
2 Months Later				0.41	- 0.31	

The influence of temperature may also be examined by relating secular change in incidence to the prevailing temperatures. Considering first the 3 months January to March, the mean quarterly air temperature was unusually low in the years 1942, 1947, and 1951, during which 22 propositi were born; the three years with the highest temperatures in this quarter were 1943, 1948, and 1950, which yielded sixteen cases. The coefficient of correlation between the number of cases in this quarter in each of the eleven years 1942-52 and mean air temperatures was -0.25. The second quarter (April-June) showed a similar difference in the number of cases between the 3 years with the lowest temperatures and the three with the highest (sixteen and ten respectively); the coefficient of correlation based on the 11 years was -0.64. The third quarter showed a slight difference in the opposite direction (seven and nine cases) and a slight positive correlation (+0.17), but in the fourth quarter the influence of temperature was again evident: fifteen propositi were born in the three coldest years and nine in the warmest, and the correlation coefficient was -0.31.

In view of the small number of observations, little importance can be attached to the magnitude of these coefficients, but it seems reasonable to suppose that the sign has some significance. The negative correlation between incidence and air temperature during the winter, spring, and autumn seasons suggests that low temperature may be of aetiological

importance. The trivial positive correlation during the warmest quarter of the year (July to September) is not necessarily inconsistent with this view, since the temperature at that time may rarely fall low enough to have any influence.

There was some indication that males were more sensitive to seasonal influences than females. Of the 27 males in the series, twenty (74 per cent.) were born in the period from October to March; the corresponding figure for females was 94 out of 159 (59 per cent.). The relative frequency of bilateral and unilateral dislocations did not appear to vary according to season of birth.

FAMILIAL INCIDENCE

- (a) Twins.—There were two male and two female propositi who were twins, all from like-sexed pairs. Dislocation of the hip was diagnosed in only one of each pair, but one unaffected twin (female) showed radiographic evidence of acetabular dysplasia. An unaffected member of one of the male pairs was reported to have talipes. The large series of twins reported by Idelberger (1951) did not show an unusual incidence of defects in the unaffected members.
- (b) Younger Sibs.—In the case of families containing two or more affected children, later sibs were enumerated from the first propositus and not from the first affected child if this was not a propositus. After excluding children who had died or were less than 3 years old at the time of the survey, there were 25 younger brothers, none of whom was affected, and 37 younger sisters of whom three were affected (Table XII). These figures are too small to permit more than a rough evaluation of the chance of a subsequent child being affected, but they suggest that the average risk may be about 5 per cent. The actual risk depends, of course, on the sex of the child; possible values are 1 to 2 per cent. for boys and 5 to 10 per cent. for girls. These estimates may vary with season of birth. Vogel's and Isigkeit's data suggest similar risks to siblings of affected. An estimate of 17 per cent. (Reed, 1955) appears to be too high.
- (c) Older Sibs.—There were 81 older brothers, one of whom had congenital dislocation of the hip, and 79 older sisters, seven of whom were affected. The incidence in prior sibs was, therefore, very similar to that in subsequent sibs.
- (d) Parents.—There were five affected mothers. No fathers were affected. No history of consanguineous marriage was obtained. These data do not, of course, permit an assessment of the risk to children of an affected parent, but if it is assumed

that the incidence has not changed and that the fertility of affected persons is not grossly impaired, it seems likely that this risk is not greater than that in children born after an affected sibling.

(e) Other Relatives.—Inquiry was made into the occurrence of congenital dislocation of the hip among siblings of the parents (uncles and aunts of propositi) and among their children (first cousins of propositi). A few mothers had only a vague knowledge of their nephews and nieces and even less of their husband's relatives. In these circumstances estimates of incidence might be liable to bias and a comparison of incidence between paternal and maternal relatives might give misleading results. We have attempted to overcome this difficulty by considering only those relatives whom the mother could name, since it seems likely that if she knew the name she would know whether or not the individual had a dislocation. Children who had died in infancy and those too young to manifest the condition were excluded. Care was taken to avoid duplication of an affected relationship; for example, if there were two propositi who were cousins only one was counted as an affected relative. Results are summarized in Table XII (overleaf).

Because of the low frequency of the condition in relatives, reliable figures of incidence cannot be obtained from a series of this size, but some broad conclusions are possible. Considering all relatives together, incidence does not appear to vary with the sex of the propositus. As expected, female relatives are affected much more frequently than males, and the proportion of males (9 per cent.) is not very different from the usual sex ratio. The incidence is considerably greater among sibs than among parents, and among cousins than among uncles and aunts. There is no indication that the disability is transmitted more frequently through the distaff side of the family.

The incidence of other malformations among relatives is of considerable interest. It was considered that adequate details of these defects among uncles, aunts, and cousins of propositi could not be expected, and the examination has been restricted to incidence among siblings. Excluding children who had congenital dislocation of the hip, there were 250 live-born siblings, of whom nine (3·6 per cent.) had congenital defects (Table XIII, overleaf). This incidence is greater than that for a representative series of live births (2·9 per cent.) reported by Grundy and Lewis-Faning (1957). The difference appears to be due to the prevalence of defects which are sometimes associated with congenital dislocation of the hip (talipes, torticollis, and hernia).

TABLE XII								
INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP AMONG RELATIVES OF PROP	OSITI							

	Relatives							
Propositi	Deletionship to December	Male		Female		Both Sexes		
	Relationship to Propositus	Total No.	No. Affected	Total No.	No. Affected	Incidence per 1,000		
Male	Younger Sibs Older Sibs Parents Uncles or Aunts (Paternal) Uncles or Aunts (Maternal) Cousins (Paternal) Cousins (Maternal)	5 9 25 46 48 22 66	0 0 0 0 0	4 10 25 40 51 33 55	0 1 2 0 0 0	7		
Female	Younger Sibs Older Sibs Older Sibs Parents Uncles or Aunts (Paternal) Uncles or Aunts (Maternal) Cousins (Paternal) Cousins (Maternal)	20 72 139 223 232 174 239	0 1 0 0 1 0	33 69 139 201 251 186 250	3 6 3 1 0 2 2	9		
Both Sexes	Younger Sibs Older Sibs Older Sibs Parents Uncles or Aunts (Paternal) Uncles or Aunts (Maternal) Cousins (Paternal) Cousins (Maternal)	Incidence per 1,000 0 12 0 0 4 0 2		0 81 12 89 0 30 4 4 0 0 9 0 7		48 50 15 2 2 2 5 3		

TABLE XIII

FREQUENCY OF MALFORMATIONS AMONG 250
LIVE-BORN SIBLINGS (EXCLUDING CASES OF
CONGENITAL DISLOCATION OF THE HIP)

Malformation	No.	Per cent.		
Talipes Torticollis Diaphragmatic Hernia		::	2 1 1	1.6
Spina Bifida Hare-lip and Cleft Palate Pyloric Stenosis Hydrocele Polydactyly			1 1 1 1 1	2.0
Total	••	••	9	3.6

DISCUSSION

The term "congenital dislocation of the hip" has continued since it was originated by Hippocrates, although its literal meaning is misleading. "Congenital" ($\dot{\epsilon}_{x}$ $\gamma\epsilon\nu\epsilon\eta\varsigma$) is inappropriate, in that the dislocation itself is probably present at birth in only a minority of cases, and "dislocation" ($\dot{\epsilon}_{x}$ $\dot{\epsilon}_{y}$ $\dot{$

Before attempting to formulate a hypothesis it may be useful to recapitulate the more important facts based on evidence derived from this and earlier inquiries:

- (1) There is a marked geographical variation in incidence.
- (2) The condition is much more frequent among females than males.
- (3) The left hip is involved more frequently than the right.
- (4) Associated defects are common. They usually involve the musculo-skeletal or nervous systems. These musculo-skeletal defects are also more frequent among siblings.
- (5) The defect occurs more commonly among first-born than among later-born, and is often associated with breech delivery.
- (6) Incidence is greater in children born in winter than in those born in summer and seems to be inversely related to air temperature.
- (7) There is a marked tendency for the condition to run in families. Maternal and paternal relatives appear equally liable to the defect.
- (8) Twin studies indicate that the foetal genotype considerably influences but does not necessarily determine the development of dislocation.

The evidence strongly suggests that both genetical and environmental factors are important. The circumstances after conception which comprise the environment may conveniently be divided into those operating before birth, during birth, and after birth, but it is not always possible to decide with certainty when a particular factor operated. As knowledge of the environment before birth is largely based on observations made at birth, and as the contribution of the foetal genotype to such circumstances as foetal lie and quantity of liquor amnii is unknown, it is difficult to obtain any information implicating the pre-natal environment which cannot be interpreted as evidence relating either to genetic factors or to parturition. But there is no firm evidence that the actual conditions of birth have any influence, and this view is supported by the observation that incidence is not reduced in children delivered by Caesarean section.

The greatly increased risk associated with primogeniture may reasonably be accepted as evidence of some features of the uterine environment present at first, but not at later, births which are sufficient to double the average liability to dislocation.

The considerable variation in liability to dislocation with season of birth suggests that the differences between summer and winter in England may be sufficient to lead to a doubling of the risk, and even greater seasonal variations have been observed in Japan (Mizuno, 1957). In attempting to explain this seasonal effect, we may first consider the possibility that it is due to variations in maternal diet. There is no doubt that there are seasonal changes in the character of the diet and particularly in its vitamin content, but it seems unlikely that this can account for the seasonal incidence of congenital dislocation of the hip as there is no evidence that the condition is commoner among malnourished populations or is associated with deficiency diseases. Another possibility is that the association may be due to the effect of temperature on the tone of the uterus and abdominal muscles. We know of no direct evidence that temperature influences the tone of the uterine musculature and it is likely that its effect on the tone of the abdominal muscles is trivial compared with the effect of previous pregnancies. The fact that the influence of season on incidence is as marked as that of parity suggests, therefore, that this explanation is not adequate. Lastly, we may consider factors in the post-natal environment which may account for this variation of incidence with season. The influence of cold in increasing the muscle tone of the infant can probably be dismissed because it would have, if anything, a protective action. The most likely explanation is that infants born in winter, particularly during cold weather, have to endure heavier clothing and cot coverings. This is likely to cause severe restriction of limb movement and prevent the thighs from assuming the natural position of flexion, abduction, and external rotation which is similar to the position adopted in the conventional treatment of the deformity. Support for this view is provided by the extremely high incidence reported among some Red Indian and Lapp communities; this may be related, in part, to the constrictions imposed by the shoe-like Red Indian cradle into which the infant is laced, and the coffin-like Lappic cot.

Evidence relating to the influence exerted by the environment after birth has not previously been seriously considered and may be received with scepticism. It is, however, no more surprising that variation in the infant's environment should influence any predisposition to dislocation than that the environment imposed by the surgeon's plaster spica should be effective in reversing this process.

We may now consider the mechanism by which genetic and environmental agencies may predispose to dislocation. This may be brought about by:

- (1) Variations in obliquity, eccentricity, or depth of the acetabulum.
- (2) Variations in the forces of approximation between the head of the femur and the acetabulum; these will be determined mainly by the muscle tone of the foetus and infant.
- (3) Variations in the other forces acting on the joint surfaces. These will be determined mainly by the position of the legs, and by the compressive forces acting on the legs of the foetus or infant, either from the uterine wall before birth or from clothing after birth.

These three groups may, of course, interact with one another. A possible form of interaction, advanced by Browne (1957), is that the application of pressure between the head of the femur and the acetabular rim may sometimes be so great as to interfere with normal growth, leading to a selfperpetuating condition of acetabular dysplasia. This concept of pressure dysplasia had previously been advanced in relation to infantile scoliosis and club foot (Browne, 1956) and probably has wide application. It is more readily appreciated if it is realized that the distance through which dislocation occurs is small compared to the change in size of the parts with growth. Although a dislocation cannot develop de novo, since the whole hip joint evolves from a single mass of cartilage, it could easily develop as a consequence of differential growth of the various parts without stretching or rupture of any part.

Evidence bearing on these points is provided by consideration of abnormalities associated with congenital dislocation of the hip. They may be classified into six groups:

- (1) Conditions leading to gross neuro-muscular disturbances. Spina bifida, amyotonia, arthrogry-posis, and cerebral diplegia are not infrequently associated with congenital dislocation of the hip. Spina bifida is a particularly common precursor, although the association will usually escape notice unless the hip joints are opened at autopsy.
- (2) Conditions predisposed to by a shallow, oblique, or eccentric acetabulum. Perthes' disease and osteo-arthritis of the hip are often associated with these variations from the normal, and the incidence of both is raised among relatives of persons with congenital dislocation of the hip (Isigkeit, 1928).
- (3) Conditions leading to generalized or localized impairment in muscle tone or co-ordination. These conditions might be expected to result in club-foot, torticollis, scoliosis, and herniae, all of which are found more frequently than usual, not only in persons with congenital dislocation of the hip (Poli, 1937) but also in their siblings and other relatives (Storck, 1938; Isigkeit, 1928). Such muscular weakness or incoordination might also predispose to inability of the foetus to assume a vertex presentation, either through inactivity or through failure to fold the legs (Vartan, 1940).
- (4) Conditions predisposed to by increased uterine pressure. Increased uterine pressure could explain most, if not all, of the associated abnormalities considered in the previous group, and also the tendency to breech delivery (Vartan, 1950). The high incidence of such abnormalities in paternal relatives and the results of twin studies limit the adequacy of this explanation. Browne (1957) considers that there is a distinct sub-group of cases of hip dislocation associated with club foot which is readily explained by extreme uterine pressure. Our data on nine cases with these combined abnormalities do not show any remarkable distribution by birth rank or method of delivery. Increase in uterine tone might also account for the impaired rate of foetal growth and perhaps for the increased incidence of toxaemia noted in this series.
- (5) Conditions predisposed to by persistent breech presentation. Torticollis appears to be the only other common malformation to which breech delivery particularly predisposes (Storck, 1950). We were not able to confirm Storck's view that a pronounced tendency to persistent breech presentation is genetically determined and is mainly responsible for the hereditary tendency to congenital dislocation

of the hip. Our data on breech delivery in siblings did not suggest that it was more frequent than in the general population. The Julio-Claudian family provides an interesting example of a familial association of obstetric difficulties, including breech delivery, and lameness.

(6) Other congenital abnormalities. These do not appear to occur with increased frequency among children with congenital dislocation of the hip and the association may be regarded as fortuitous. Poli's observations are consistent with this interpretation of our more limited data.

Any attempt to postulate an acceptable genetic hypothesis for congenital dislocation of the hip must be made in cognizance of the fact that environmental variations may greatly increase the liability of manifestation, and that the hip is a structure of such variability and complexity that it would seem wholly unrealistic to expect that such a deformity should be determined by a single gene substitution. The fact that the pelvis shows appreciable sexual differences, even as early as the third month of foetal life (Dega, 1933) suggests that it is not necessary to postulate sex-linked genes.

Because of the large number of genetical and environmental factors which appear to influence the liability of the hip to dislocate, the degree of stability of the articulation in a population of new-born infants would be expected to show continuous variation. The imagined distribution of a series of hip joints is shown in Fig. 2 (opposite), the more stable joints being on the left and the more labile on the right.

Let us suppose that such a series is exposed to varying degrees of deforming force, represented by the dotted line in the figure. Joints indicated by the area ABB' would never dislocate, however unfavourable the postnatal environment. Those distributed near to the line CC' would be dislocated only by very unfavourable circumstances, and those near to the line DD' would usually dislocate. The most labile joints, represented by the area EFE', would always dislocate, however gently the infant were handled, and would frequently be dislocated before birth.

The orientation of the frequency distribution relative to the horizontal axis depends on a number of factors. There is evidence that, because of differences in the depth and inclination of the acetabulum, the hip joints of Sudanese and possibly of most indigenous African peoples are stronger than those of white races (Cheynel and Huet, 1952).

Fig. 3 (opposite) shows hypothetical distributions of the hip joints of two populations, the solid line

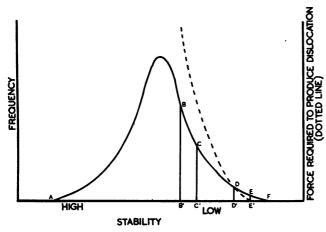


Fig. 2.—Hypothetical distribution of hip-joints according to stability.

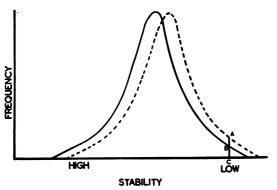


Fig. 3.—Hypothetical distribution of hip-joints of two populations according to stability.

representing an African and the dotted line a white population. The application of a given stimulus (ABC) to the joints of both groups would clearly cause more dislocations among the whites than among the Africans.

The same sort of model may be used to account for the differences in incidence between males and females (males being represented by the solid line and females by the dotted line), and between the two sides (solid line for the right side and dotted line for the left). These differences are supported by observations based on measurements of the hip joints of one hundred foetuses reported by Dega (1933).

It appears, then, that the hypothesis examined here is not inconsistent with the more important points known about congenital dislocation of the hip. It is possible that the general terms of the hypothesis are more widely applicable and may help in understanding other defects. We have, at present, no conclusive evidence that this is true of other human malformations, but there are indications that some animal abnormalities can be explained in this way. Grüneberg (1951) has described a foramen in the acetabulum of the mouse apparently determined by multifactorial inheritance and showing what he terms quasi-continuous variation. Other abnormalities in mice inherited in a similar way have been shown to be influenced by the uterine environment (Deol and Truslove, 1957). The crookedtoe defect of poultry is considered by Hicks (1953) and Lerner (1954) to be the expression of one tail of a continuous distribution determined by multifactorial inheritance. Manifestations of the condition is influenced by sex and by such environmental factors as walking on wire-netting.

Lerner also considers that certain malformations of the wings of the fruit fly investigated by Goldschmidt (1945) and Goldschmidt, Hannah, and Piternick (1951) can be explained in a similar way. All these malformations are associated with impairment in fertility.

The hypothesis may be criticized, as other polygenic theories have been, on the grounds that it cannot fail to agree with all observations relating to the inheritance of the disease. Nevertheless, it seems to be more realistic than hypotheses which involve only one or two genes. It may also be of more practical value because it does not divert attention from environmental influences. In the present state of knowledge it is questionable whether eugenic counselling is justifiable, and it would probably be more useful to attempt to control environmental influences by the early recognition and rectification of foetal malpositions and by permitting the infant full limb movement and the adoption of a natural position. It seems quite certain that the advising of parents with an affected child not to reproduce, even if the advice were acted upon, would have a negligible influence on the incidence of the disease. Affected parents do not appear to be responsible for more than about 5 per cent. of cases. A form of advice which would be at least harmless, and might reduce the risks by as much as a half, would be to suggest the late autumn as the most propitious time for conception.

SUMMARY

(1) An attempt was made to ascertain all children who were born in the period 1942-52 and were treated in Birmingham hospitals for congenital dislocation of the hip. The incidence was 0.65 per

1,000 live births. Further information on 167 patients was obtained by interviewing the mothers.

- (2) The data were consistent with previous reports relating to the sex ratio (15 per cent. males) and the side more commonly affected (64 per cent. of affected hips were left-sided). The frequencies of associated defects (16 per cent.), and of breech delivery (17 per cent.) were rather higher than in most previous reports.
- (3) The mean birth weight was about $\frac{1}{2}$ lb. below the expected value. This could not be explained by any obvious decrease in the duration of gestation.
- (4) The incidence among first-born was about twice that in second and third children. There appeared to be some association with increasing maternal age, and there was some evidence that the fertility of mothers was slightly impaired.
- (5) The incidence among children born in the winter was double that of children born in the summer. Examination of annual variations suggested that incidence was related to air temperature.
- (6) The risk of the condition occurring in a subsequent child appears to be about 5 per cent., but is dependent on the sex of the child and possibly on the season of birth.
- (7) Current hypotheses are considered in relation to these findings. It is suggested that the stability of the hip joint is determined by multifactorial inheritance and by a number of factors in the pre-natal and post-natal environment. The most important of these are considered to be those which influence the attitude and activity of the foetus and of the infant.

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REFERENCES Allison, A. C. (1956). Personal Communication.
Browne, D. (1956). Proc. roy. Soc. Med., 49, 395.
— (1957). Personal Communication.
Cheynel, J., and Huet, R. (1952). Rev. Orthop., 38, 279.
Dega, W. (1933). Chir. Organi. Mov., 18, 425.
le Damany, P. (1912). "La luxation congénitale de la hanche".
Alcan, Paris. Alcan, Paris.
— (1914). Amer. J. orthop. Surg., 11, 541.
Deol, M. S., and Truslove, G. M. (1957). J. Genet., 55, 288.
Faber, A. (1937). Z. Orthop., 66, 140.
Getz, B. (1955). Acta orthop. scand., Suppl. 18.
Gibson, J. R., and McKeown, T. (1950). Brit. J. soc. Med., 4, 221.
Goldschmidt, R. B. (1945). J. Morph., 77, 71.
—, Hannah, A., and Piternick, L. K. (1951). Univ. Calif. Publ.
Zool., 55, 67.
Grundy E. and Lewis-Enning F. (1957). "Mochidity and Mortality.

Zool., 55, 67.
Grundy, F., and Lewis-Faning, E. (1957). "Morbidity and Mortality in the First Year of Life". Eugenics Society, London.
Grüneberg, H. (1952). J. Genet., 51, 95.
Hatt, V. L. (1947). Minn. Med., 30, 889.
Hicks, A. F., Jr. (1953). "The Genetics and Development of the Crooked-toes Defect in Chickens". Ph.D. Thesis, Univ. California.
Idelberger, K. (1951). "Die Erbpathologie der sogenannten angeborenen Hüftverrenkung". (Suppl. to Beitr. klin. Chir.) Urban and Schwartzenberg. Munich.

Schwartzenberg, Munich.

Isigkeit, E. (1928). Arch. orthop. Unfall-Chir., 26, 683.

Lerner, I. M. (1954). "Genetic Homeostasis". Oliver and Boyd, Edinburgh.

MacMahon, B., Record, R. G., and McKeown, T. (1951). Brit. J. soc. Med., 5, 185.

McKeown, T., MacMahon, B., and Record, R. G. (1951a). Ann. Eugen. (Lond.), 16, 249.

—, —, —, (1951b). Lancet, 2, 556.
—, —, —, (1952). Arch. Dis. Childh, 27, 386

Philadelphia

Philadelphia.
Sainton, R. (1893). Rev. Orthop., 4, 352.
Smith, A., and Record, R. G. (1955). Brit. J. prev. soc. Med. 9, 51.
Steinberg, A. G. (1957). Personal Communication.
Storck, H. (1938). Z. Orthop., 68, 308.
— (1950). Ibid., 79, 282.
Vartan, C. K. (1940). Lancet, 1, 595.
— (1950). In "Modern Trends in Obstetrics and Gynaecology", ed. K. Bowes, p. 196. Butterworth, London.
Vogel, K. (1905). Z. orthop. Chir., 14, 132.